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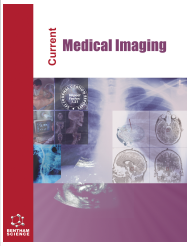

CASE REPORT

Fibrosarcoma of Great Toe with Multimodality Radiologic Imaging Features

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Abstract:

Introduction:

Fibrosarcoma of bone is a rare malignant spindle cell tumor.

Case Study:

Herein, we present a case of fibrosarcoma in a 40-year-old male who was presented to the clinic with left-sided great toe pain for 20 years. Simple radiographs showed acrolysis at the distal phalanx of great toe. Magnetic resonance imaging (MRI) revealed a 1.5 cm sized heterogenous high signal intensity mass on T2-weighted images and iso signal intensity on T1- weighted images. Dorsal and distal portion of the mass showed markedly dark signal intensity on T1 and T2-weighted images.

Conclusion:

In an enhanced image, the mass showed heterogenous enhancement. Surgical removal was performed and pathologic analysis revealed fibrosarcoma. Although extremely rare, fibrosarcoma of the bone should be kept in mind as a possibility when a lesion exhibits a black signal intensity component on an MRI with acrolysis.

Keywords: Fibrosarcoma, Hallux, Phalanx, Magnetic resonance imaging, Radiographs, T2-weighted.

Article History	Received: December 27, 2022	Revised: May 16, 2023	Accepted: May 24, 2023

1. INTRODUCTION

Fibrosarcoma of bone is a spindle cell malignant neoplasm which is composed of malignant spindle-shaped fibroblasts with collagen production. It is very rare and can be diagnosed when other diagnoses are excluded [1]. Fibrosarcoma usually involves deep soft tissue and is mostly located in the trunk and the extremities. Primary fibrosarcoma of bone is a rare tumor, accounting for less than 5% of bone sarcomas [2 - 4]. If it occurs in the bone, fibrosarcoma is usually located in the proximal and distal femur, proximal tibia, and distal humerus. Clinically, patients represent local pain, swelling, and limitation of motion [1]. Fibrosarcoma of bone shows relatively uniform incidence from the second to sixth decades of life [5]. Regarding treatment, wide local excision is the first-line treatment option, and radiotherapy/chemotherapy can be used in patients with unresectable, recurrent, or metastatic cases [6].

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Herein, we present a case of fibrosarcoma in a 40-year-old man with acrolysis at a distal phalanx of the great toe. The purpose of this study is to describe radiologic features including radiography and magnetic resonance imaging (MRI), and pathologic features of fibrosarcoma of bone.

2. CASE REPORT

This study was approved by the Institutional Review Board of our hospital and the requirement for informed consent was waived because it is retrospective study based on the medial record.

A 40-year-old male presented with spontaneous great toe pain with an ingrown nail over 20 years. He came to our outpatient orthopedic clinic because of aggravating sharp pain [Score 3 according to numerical rating scale between 0 and 10] with palpability. On physical examination, a focal swelling of the great toe tip was noted without nail deformity or skin color change. There was no history of antecedent surgery or trauma.

Initial radiographs showed acrolysis at the distal phalanx of the great toe. The distal end of remnant epiphysis of the distal

phalanx had a sclerotic margin. There was no discernible underlying mineralization nor periosteal reaction on the great toe tip (Fig. **1A**). CT scan showed iso-attenuating mass with acrolysis of the distal phalanx and clear sclerotic margin. On SPECT CT, the lesion showed a cold defect in the distal phalangeal portion of the great toe (Fig. **1B**). From these radiographic findings, we considered this lesion as benign lesion such as ganglion cyst or low-grade malignant lesion. However, foot MRI (Fig. **1C**) revealed 1.5 cm sized heterogenous high signal intensity mass on T2- weighted images and iso signal intensity mass on T1- weighted images. Dorsal and distal portion of the mass showed markedly dark signal intensity on T1 and T2WI representing fibrotic component. The margin of the mass showed speculation with a desmoplastic reaction to adjacent tissue. After administration of gadolinium, the lesion showed peripheral and heterogenous enhancement on fat-suppressed postcontrast T1- weighted images. High b-value (b=1000) diffusion-weighted image showed restriction of the lesion with low ADC (Apparent Diffusion Coefficient) value. Combining MRI findings, we made the final radiologic differential diagnoses as giant cell tumor of the tendon sheath and glomus tumor.

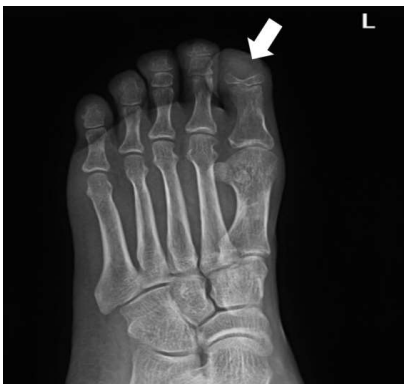

Fig. (1A). Initial radiograph showed acrolysis (arrow) at the left great toe distal phalanx.

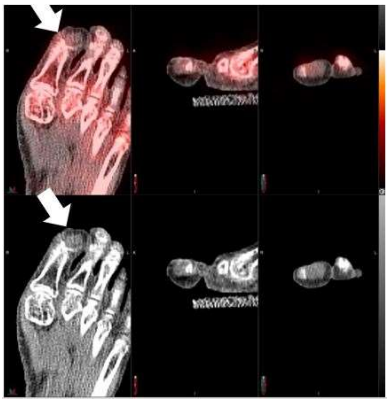

Fig. (1B). SPECT CT.
On SPET CT scan, lesion (arrows) showed iso-attenuating mass and cold defect in the distal phalangeal portion of great toe.

He underwent a surgical excisional biopsy. The tumor was composed of relatively uniform spindle cells arranged in sweeping fascicles with a herringbone pattern. The tumor cells showed mild nuclear atypia with small inconspicuous nucleoli. Mitotic activity was not identified (Fig. **1D**). By immunohistochemistry, the tumor cells showed focally expressed smooth muscle actin and Ki-67 labelling index was very low, less than 2%. Desmin, S-100 protein and CD99 was negative. According to the typical histologic findings, the tumor was diagnosed as fibrosarcoma.

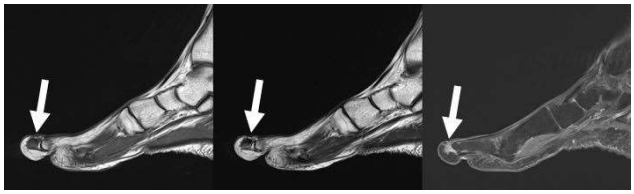

Fig. (1C). MRI.
Sagittal T1WI (left), T2WI (middle) and CE-T1WI (right) show 1.5 cm sized mass (arrows) with iso signal intensity on T1WI and heterogenous high SI on T2WI. After contrast injection, lesion showed heterogenous and peripheral enhancement.

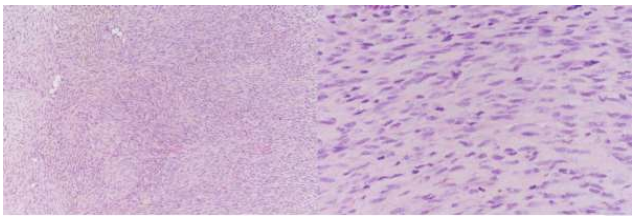

Fig. (1D). The tumor cells arranged in long fascicles with herringbone pattern (left) (HE, x100).
There is mild nuclear atypia with small nucleoli (right) (HE, x400).

3. DISCUSSION

Fibrosarcoma is a rare sarcoma composed of relatively monomorphic malignant fibroblasts with collagen production. Fibrosarcoma of bone typically includes long bones that prefers distal femur and proximal tibia [1, 7]. According to the literature, thirty to sixty percent of fibrosarcoma occur in the lower extremity, but only 2.5% are located within the foot and ankle [8]. There are two types of fibrosarcoma of bone. Primary fibrosarcoma arises in the medullary canal or periosteum. Secondary fibrosarcoma of bone arises from preexisting lesions such as skin fibrous sarcoma, solitary fibrous tumor, and dedifferentiated liposarcomas [9, 10].

Radiographic findings show a lytic and destructive lesion with cortical erosion. Our patient showed similarities representing osteolytic lesions with sclerotic margins. On MRI scans, most patients demonstrated hypointense on T1-; weighted and heterogenous signal intensity on T2-weighted images with an associated soft tissue mass [11]. Also, the lesion showed band-like areas of hypointense on both non-enhanced images. According to the literature, a hypointense band like lesion is thought to result from the presence of collagen fibers in the tumor stroma and suggest that tumor is fibrous [9]. In our case, the mass showed a markedly dark signal intensity band-like area on T1 and T2-weighted images, representing fibrous component consistent with previous findings. After contrast injection, lesion showed mild enhancement [11]. Regarding DWI, Wang's study reported two cases of soft tissue fibrosarcoma performed DWI and lesions showed diffusion restriction like our patient. Diffusion

restriction indicated dense cellularity of the tumor [9].

In retrospect, fibrosarcoma can be considered if it is a tumor that lyses the bone showing the characteristic of a malignant tumor and also has abundant fibrous tissue.

The differential diagnosis may include desmoplastic fibroma, intraosseous epidermal inclusion cyst, and giant cell tumor. Desmoplastic fibroma usually involves the mandible, large long bone or iliac bone. On a radiograph, desmoplastic fibroma represents well-defined non-sclerotic marginated geographic lesion. The cortex is usually thinned and cortical breakage is common. On MRI, lesion shows intermediate to hypointensity on T1-weighted and T2-weighted images with heterogenous enhancement or rim enhancement on contrast enhanced fat suppressed T1-weighted image [12, 13]. Intraosseous epidermal inclusion cyst manifests as well-defined expansile osteolytic lesion on radiograph and typically the bony reaction is absent [14]. On MRI, lesion showed intermediate signal intensity on T1 and T2-weighted images, and after gadolinium administration, the lesion showed peripheral rim enhancement. These MRI findings are not compatible with fibrosarcoma. Giant cell tumors are commonly seen in the distal femur, proximal tibia, and distal radius and giant cell tumor of bone is known to be rare in the phalanx of the foot [12, 15]. Imaging characteristic of giant cell tumor is nonspecific and shows low to intermediate signal intensity at both T1 and and T2-weighted image [16].

Treatment options include limited excision, wide local excision, amputation, and local excision with adjuvant treatments [5].

Previous study reported that 15% of patients had local recurrence after treatment. Also, metastasis developed in 68% of patients, most commonly in the lung and skeleton [2].

Imaging studies in patients with fibrosarcoma of bone are very rare and we hope this report can be helpful for radiologists to diagnose the fibrosarcoma.

CONCLUSION

In conclusion, we have presented a rare case of fibrosarcoma with acrolysis of the great toe. Clinical presentation and imaging features are unspecific, and if a malignant tumor with dark signal intensity is suspected, it is necessary to consider fibrosarcoma as a differential diagnosis.

LIST OF ABBREVIATIONS

MRI = Magnetic Resonance Imaging

ADC = Apparent Diffusion Coefficient

T2WI = T2 Weighted Image

SPECT = Single-photon Emission Computed Tomography

ETHICAL STATEMENT

This study was approved by the Institutional Review Board of our hospital.

HUMAN AND ANIMAL RIGHTS

Not applicable.

CONSENT FOR PUBLICATION

The requirement for informed consent was waived because it is retrospective study based on the medial record.

STANDARDS OF REPORTING

CARE guidelines and methodology were followed.

AVAILABILITY OF DATA AND MATERIALS

The data and supportive information are available within the article.

FUNDING

This study was supported by Soonchunhyang Research fund.

CONFLICT OF INTEREST

The authors declare no conflict of interest financial or otherwise.

ACKNOWLEDGEMENTS

Declared none.

SUPPLEMENTARY

MR parameters.

The parameters of sagittal T1 TSE sequence were as follows: repetition time / echo time, 632 / 11; matrix, 512 × 333; echo train length, 1; number of signal acquisitions, 1; field of view, 24 cm; section thickness, 3 mm; and interslice gap, 0.3 mm. The parameters of sagittal T2-weighted TSE sequence was as follows: repetition time / echo time, 5750 / 92; matrix, 512 × 358; echo train length, 1; number of signal acquisitions, 1; field of view, 24 cm; section thickness, 3 mm; and interslice gap, 0.3 mm. The parameters of sagittal T1 fat-suppressed contrast enhancement sequence was as follows: repetition time / echo time, 610 / 11; matrix, 512 × 333; echo train length, 1; number of signal acquisitions, 1; field of view, 24 cm; section thickness, 3 mm; and interslice gap, 0.3 mm; contrast agent, Gadovist(gadobutrol, 1.0mmol/ml, 10cc, bolus). (3.0-T MRI, Magnetom Skyra; Siemens Healthcare, Erlagen, Germany).

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